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Takayasu Arteritis Associated with Ulcerative Colitis: Literature Review of HLA Profile

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Abstract **Case Report**

Takayasu arteritis and ulcerative colitis are two chronic inflammatory diseases of unknown etiology and their coexistence is very rare. In this report, we describe a case of 51 years-old Moroccan man with a 4 years history of ulcerative colitis confirmed with pathologic examination who developed Takayasu arteritis. The patient was admitted several times for recurrent arterial and venous thrombosis; during the etiologic investigation, Takayasu's arteritis was diagnosed. The coexistence of these two pathologies is a rare entity, reported accidentally in the world especially in Southeast Asia. This association supposed common pathophysiological mechanisms. Different assumptions, genetic factors and environmental agents that contribute to the pathogenesis of these diseases, have to be validated by others studies to clarify these mechanisms.

Keywords: Takayasu arteritis - ulcerative colitis – Thrombosis – HLA.

INTRODUCTION

Takayasu arteritis (TA) is a rare systemic inflammatory large-vessel vasculitis of unknown etiology that most commonly affects women of childbearing age. Ulcerative colitis (UC) is an inflammatory bowel disease that causes lasting inflammation and ulcers in digestive tract. TA is very common in Japan and Southeast Asia, whereas UC is common in north America and Europe [1]. The association of these two diseases is very rare and the little cases were reported in the literature, with only some fifteen cases reported worldwide and more than half of them from Japan [1, 2]. This association has not been reported previously in Morocco. We present a case report of association of these two diseases in 51 years-old Moroccan man.

CASE PRESENTATION

A 51-year-old Moroccan male was followed since 2010 in gastroenterology department for ulcerative colitis revealed by rectorrhagia with a rectal syndrome confirmed by rectos copy and rectal biopsy with anatomopathological study, the patient was treated with sulfasalazine with a good clinical evolution. Four years later, the patient was admitted for the pulmonary embolism treated with anticoagulant therapy with a good clinical evolution. In the same year, the patient was admitted in the cardiac intensive care unit for cardiogenic shock. A thoracic computed tomography was performed urgently showed a bilateral proximal pulmonary embolism associated with thrombosis of the inferior and superior vena cava. An echocardiography was performed objectified an acute cor pulmonale with tricuspid insufficiency and pulmonary hypertension at 93 mmhg, inferior vena cava was dilated with proximal thrombosis. The patient was treated successfully with thrombolysis.

The patient was admitted in internal medicine department for etiological assessment of thromboembolic disease. Physical examination on admission revealed a body temperature at 37.4 C, asymmetric blood pressure (110/75 mmhg in the left arm; 140/90 in the right arm), systolic pressure index was at 1.1; heart rate was 82 bpm, respiratory rate was 19 cpm, weight was 75 kg, urine test strip was negative. The cardiovascular and pulmonary examination was normal. As part of the extension assessment of thromboembolic disease, a Doppler ultrasound of the vessels of the neck showed abnormal Doppler flow in the left vertebral artery without significant stenosis, while the other supra aortic trunks were normal; Abdominal computed tomography was performed revealed left iliac vein thrombosis with circumferential wall thickening of abdominal aorta suggestive of Takayasu arteritis with no abdominal-pelvic mass **(Figure 1; 2; 3)**. The routine

blood test revealed hemoglobin of 15.4 g/dl; platelet count of 122000 e/mm3; white cell count of 6240 e/mm3; lymphocyte count of 1888 e/mm3; liver test, blood ionogram, lipid profile and renal function were normal; INR was at 2.2; prothrombin ratio was 49%; CRP was 20 mg/l; the erythrocyte sedimentation rate (54 mm in the first hour); activated antithrombin was at 103%; activated protein C was at 20%; activated protein S was at 45%. Immunological test: Anti-cardiolipin antibodies IgM was positive at 34,4 UI/l; IgG was negative. The results of HLA testing class I was B51 positive. The patient was treated with antivitamin K with an INR control at 2.69; methotrexate 15mg/week was prescribed and prednisone 1 mg/kg/day, whereas sulfasalazine was maintained. The evolution was marked by stabilization of symptoms.

DISCUSSION

TA and UC are two chronic inflammatory diseases of unknown etiology. Both diseases are not very common in Morocco. TA is a chronic inflammatory vasculitis of unknown etiology that affects the aorta and its branches and the pulmonary arteries that affect predominantly women between 10 and 40 years of age in the Asian population [1-3], however, UC is a chronic inflammatory bowel disease of unknown etiology in relapsing episodes of remission, it is common in North America and Europe and in Asia and other countries. Autopsy prevalence of 33 in Japan and northern Europe [4, 5], the incidence and prevalence of this disease in Japan are respectively 0.36/100,000 in 1980 and 7.85/100,000 in 1985 [6]. Several pathologies have already been associated with TA: inflammatory colitis, rheumatic pelvispondylitis and idiopathic retroperitoneal fibrosis [4]. The association between TA and UC has been reported in the literature by Asian authors, occasionally in Japan [2, 3].

Genetic factors are assumed to play an important role in the pathogenesis of these 2 diseases. Patients with TA and UC tend to be HLA-positive: HLA A24 and B52 [3, 5, 7] also B5 and DR2. HLA typing have been reported only 23 cases until now with this association so far including 19 patients from Japan **(Table I)** [2]. HLA typing analysis of these patients showed that this association is present in 17 women, the average age of patients was 27 years. For locus A, 16 cases of 20 patients were positive for A24 or 9 (80%). For locus B, 20 cases of 23 patients were B52 positive or five (87%). For locus DR, 14 cases of 18 patients were positive for DR2 (77%) [2]. The presence of the DR2 allele is a genetic field similar to that of Asian patients where TA and UC were observed. Researchers in various previous studies have demonstrated a strong association of HLA-B52 in TA. In the study of Ziver Sahin and al [8], with a relatively large sample included 330 patients with TA. HLA class I of these patients found a significant association of HLA-B52 with TA. The distribution of HLA-B51 did not differ between TA patients and healthy controls. The presence of HLA-B52 decreased in late-onset patients (> 40 years). Patients with angiographic type I disease with limited aortic involvement also had a lower presence of HLA-B52 compared to those with all other disease subtypes. HLA-B51 as in this case is especially reported in Behçet disease.

Among patients reported by TAKAHASHI *et al.*, [2], as in our case, TA follow the occurrence of UC in 15 cases of 21 patients (71%). UC tended to occur simultaneously or prior to the development of TA. This chronological finding may suggest that bacteremia favored by intestinal mucosal injury during UC is implicated in the onset of TA in subject with genetic predisposition [4]. These hypotheses and many others must be validated by further studies and genetic tests to understand the physio pathological mechanisms between these diseases.

Inflammatory bowel disease is a risk factor of thromboembolic complications [9,11,12], as in this case; it was described at first in 1936 by Bargen and al, who reported 18 patients with thromboembolic diseases with venous predominance [12], this risk is higher in Crohn's disease than UC [9, 12, 13]. This risk is very high during activity period of the disease [10]. In the earliest studies evaluating the incidence of thromboembolic disease in chronic inflammatory bowel disease, 0.84% of patients develop thromboembolic diseases within 11 years of evolution [12].

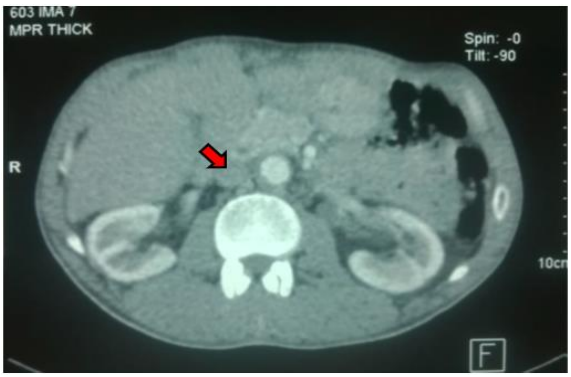

Figure 1: Inferior vena cava thrombosis (arrow)

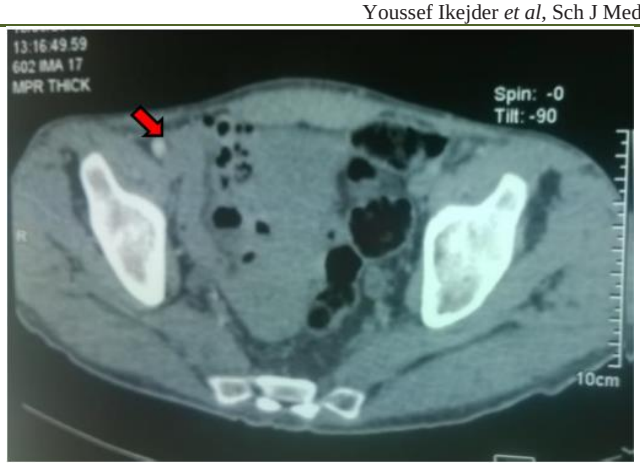

Figure 2: Left iliac vein thrombosis (arrow)

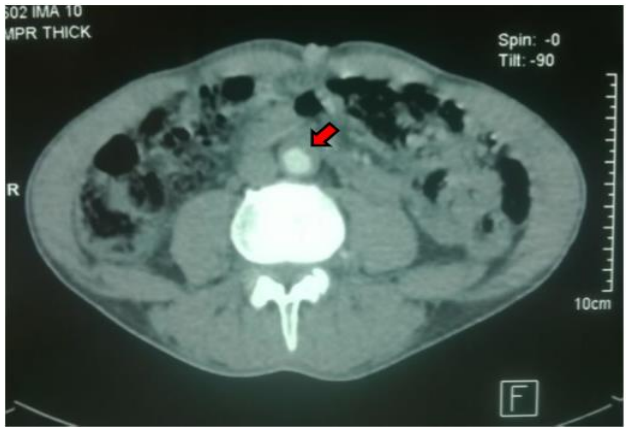

Figure 3: Wall thickening of abdominal aorta (arrow)

Table I: Review of published cases of TA associated with UC who underwent serological HLA analysis

No	Authors	Published year	Patient's age	Sex	HLA typing
1	Miwa Y	1979	43	F	A2, A9, B5, B13, Cw3
2	Achar KN	1986	35	F	A11, B5, B7, DR2, DR4
3	Ichikawa M	1988	24	F	A2, A24, B51, B52, Cw3
4	Goto M	1991	19	F	A24, A31, B52, Bw 61, Cw3
5	Yoshida H	1992	21	M	A11, A24, B52, DR2, DQm1
6	Ishikawa H	1993	27	F	A2, A24, B52, Bw 61
7	Oyanagi H	1994	25	F	A24, A33, B52, B44, DR2
8	Sato R	1994	14	F	A11, A24, B48, B52, DR2, DR9, DQ1, DQ3
9	So S	1995	27	F	A11, A24, B52, DR2, DR6
10	Morita Y	1996	19	F	A24, B51, B52, DR2, DR12
11	Aoyagi S	1998	26	M	A2, A24, B27, B67, Cw1, Cw7, DR1, DR2
12	Ito Y	2001	21	F	A24, A26, B52, B61, Cw3, DR2, DR8, DQ1
13	Suzuki T	2001	14	M	A24, A33, B52, DR2
14	Shibata C	2002	42	F	A24, A26, B35, B52, Cw3, DR2, DR 4
15	Fukunaga	2002	18	F	B52, DR2
16	Masuda H	2002	41	F	A2, A31, B52, DR2
17	Masuda H	2002	20	F	A2, A31, B52, B61, DR2, DR4
18	Bansal R	2003	15	F	A24, B15, B52, DR4, DR15
19	Hokama A	2003	36	F	A2, B35, Cw3
20	Nakano H	2004	49	M	A24, A26, B52, B4, DR51, DQ6
21	Katsinelos P	2005	36	F	B52, DR2
22	Nobuyuki T	2010	29	M	A11, A24, B52, B62, DR4, DR9
23	Current case	2015	51	M	B51

CONCLUSION

The coexistence of TA and UC is a rare entity, reported occasionally in several countries in the world especially in Japan. This association assumes the same physio pathological mechanisms and etiologies. Different hypotheses will must be validated by further studies to conform this relationship.

DECLARATION

Data Availability:

The data used to support the findings of this case report are available from the corresponding author upon request (Thoracic and abdominal computed tomography / anatomopathological study / Results of HLA testing class I /...).

Conflicts of Interest:

The authors declare that there are no conflicts of interest regarding the publication of this paper.

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